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Case Report

Osteoblastoma in the region of the hip

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Abstract

Osteoblastoma occurring in the region of the hip is very rare, and vague symptoms with uncharacteristic radiographic features often lead to misdiagnosis. Because of radiographic and histological similarities, it must be carefully distinguished from osteoid osteoma, aneurysmal bone cyst, giant cell tumor, and osteosarcoma. Computed tomography is the preferred imaging modality as it is able to detect the nidus and images will not exhibit the flare phenomenon caused by surrounding inflammation seen with magnetic resonance imaging. For hip joint lesions in weight-bearing areas, intralesional curettage may achieve satisfactory outcomes as compared with wide resection. We herein report two cases of osteoblastoma in the hip region in which diagnosis was delayed that were successfully treated with curettage and followed by high-speed burring.

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1. Introduction

Osteoblastoma is a rare, osteoid tissue-forming primary neoplasm of the bone which comprises approximately 1% of all primary bone tumors.¹ It is usually seen in adolescents and young adults, and arises within the medullary cavity or on the surface of long bones and posterior elements of the spine. Osteoblastoma in the region of the hip is rarely encountered. Osteoblastoma has many clinical and histological similarities with osteoid osteoma, but it is typically much larger in size (>1.5 cm). Clinical symptoms are vague and nonspecific, and include reduced range of motion (ROM) of adjacent joints and

nocturnal bone pain that does not respond well to nonsteroidal anti-inflammatory drug therapy.² In radiographic examinations, the initial changes are often uncharacteristic, causing further delay in diagnosis and inappropriate treatments. The treatment of choice is intralesional curettage or *en-bloc* excision, depending on the site of the lesion.³ Herein, we report two cases of osteoblastoma in the hip region.

2. Case reports

2.1. Case 1

A 13-year-old male was referred to our hospital for management of an acetabular tumor with rapid growth over the past 3 months. He had previously been seen at another hospital because of right pain hip at night with

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restricted hip motion for more than 6 months and was treated symptomatically.

At our hospital, he was noted to be limping, and physical examination revealed tenderness over the right inguinal area and limited ROM of the right hip due to pain. Plain radiographs of the pelvis showed a mixed osteolytic and osteoblastic expansile lesion over the right acetabular region with suspected periosteal reaction (Fig. 1). Whole-body bone scan showed an intense focus in the area of the right acetabulum without evidence of distant metastasis. Magnetic resonance imaging (MRI) of the pelvis showed an osteolytic bone mass $6.9 \times 6.2 \times 5.9$ cm in size at the right acetabulum, protruding into the pelvic cavity. In addition, bone marrow, soft tissue, and muscle edema in the area of the right pelvis was noted. The initial diagnosis included osteogenic sarcoma or aggressive osteoblastoma.

Needle biopsy of the lesion was performed, and histological examination was consistent with osteoblastoma (Fig. 2). Through an anterior approach, the tumor was removed by completed intralesional curettage and high-speed burring followed by cauterization with phenol. Postoperatively, touch weight-bearing was allowed for 3 months, followed by partial weight-bearing for 6 weeks, and then full weight-bearing.

Thereafter, the patient was able to walk without support, and the Harris Hip Score was 100 when followed-up at 38 months. Follow-up imaging studies of the pelvis 3 years after surgery revealed no evidence of recurrence (Fig. 3).

2.2. Case 2

A 14-year-old male was originally seen at another hospital with restricted motion of the right hip and pain for more than 3 months after a contusion injury. He received a synovectomy for presumed septic arthritis; however, histological examination of the surgical specimen revealed a diffuse-type tenosynovial giant cell tumor. He was referred to our hospital for further management.

Radiographs of the pelvis showed an osteoblastic lesion over the right greater trochanter (Fig. 4). Whole-body bone scan showed an intense focus in the area of the right greater trochanter, with increased soft-tissue activity in the lateral aspect of the right hip. MRI of the pelvis revealed a tumor growing outward from the right greater trochanter and femoral neck, and postoperative changes including scarring due to the previous lateral approach, soft-tissue edema at the right *psoas* and *gluteus* muscles and neighboring fascia, bone marrow edema at the right femoral neck and head, and hip joint effusion. Aggressive osteoblastoma, pigmented villonodular synovitis, and recurrent giant cell tumor were considered.

The lesion was removed by intralesional curettage and high-speed burring followed by chemical cauterization. Histological examination of the surgical specimen was consistent with osteoblastoma (Fig. 5). Postoperatively, touch weight-bearing was allowed for 3 months followed by partial weight-bearing for 6 weeks, and then full weight-bearing.

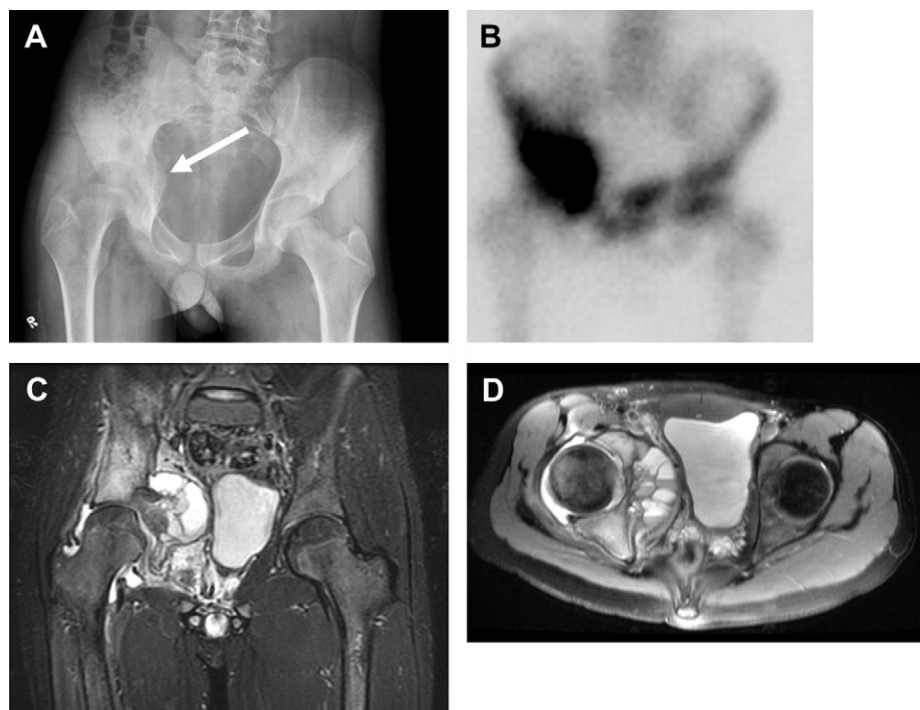


Fig. 1. (A) Anteroposterior radiograph of the pelvis revealed a mixed osteolytic and osteoblastic expansile lesion in the right acetabular region with suspected periosteal reaction. (B) Whole-body bone scan showed a marked intense focus at the right acetabulum. (C) Coronal and (D) axial pelvic magnetic resonance imaging revealed a $6.9 \times 6.2 \times 5.9$ -cm osteolytic bone mass at the right acetabulum protruding into the pelvic cavity, with internal hemorrhage and periosteal reaction at the margin of the mass. Bone marrow and surrounding soft tissue edema were present.

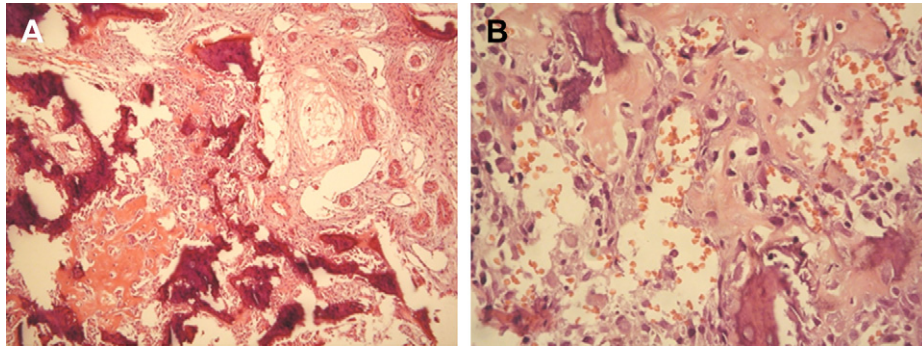


Fig. 2. (A) Histologic specimen showed interlacing net of osteoid trabeculae made of woven bone (hematoxylin and eosin; original magnification $\times 40$), which was (B) surrounded predominantly by osteoblasts and some spindle cells, giant cells or blood vessels (hematoxylin and eosin; original magnification $\times 200$).

At 7 months postoperatively, the patient was able to walk without support, and the Harris Hip Score was 100. Radiography 23 months postoperatively revealed no evidence of recurrence (Fig. 5).

3. Discussion

Osteoblastoma is a rare bone tumor affecting the long bones and posterior elements of the spine, and accounts for 1% of primary bone tumors. Osteoblastoma in the region of the hip is exceedingly rare, accounting for 3–8.8% of all osteoblastomas.^{1,4} Due to the non-specific clinical features and low incidence of the tumor in the hip, the diagnosis is often missed or delayed.

Data of seven cases of osteoblastoma in the hip region, five in the literature and the two presented herein, are summarized in Table 1.^{3,5–8} The average interval from symptom onset to correct diagnosis was around 1 year (range 6–36 months).

The initial diagnoses were varied and included septic arthritis, osteosarcoma, giant cell tumor, Legg-Calvé-Perthes disease, synovitis, enteropathic arthritis, and sacroilitis. Thus, patients often initially received inappropriate treatments such as antibiotics, synovectomy, and debridement. The various initial diagnoses illustrate the difficulty of arriving at a correct diagnosis based on initial presentation and radiographs.

Radiographic diagnosis of lesions in the region of the hip is even more challenging due to the complex anatomy of the pelvis. McLeod et al⁹ reviewed 123 cases of osteoblastoma and stated that radiographically, most tumors are within an intact cortex, usually expanded and thinned, with prominent periosteal new bone formation and various degrees of internal ossification. The feature of cortical expansion may make osteoblastomas appear similar to an aneurysmal bone cyst or giant cell tumor; however, no central calcification should be found within the latter tumors.¹⁰ Osteoid osteomas may also appear similar on radiographs; however, they are usually much

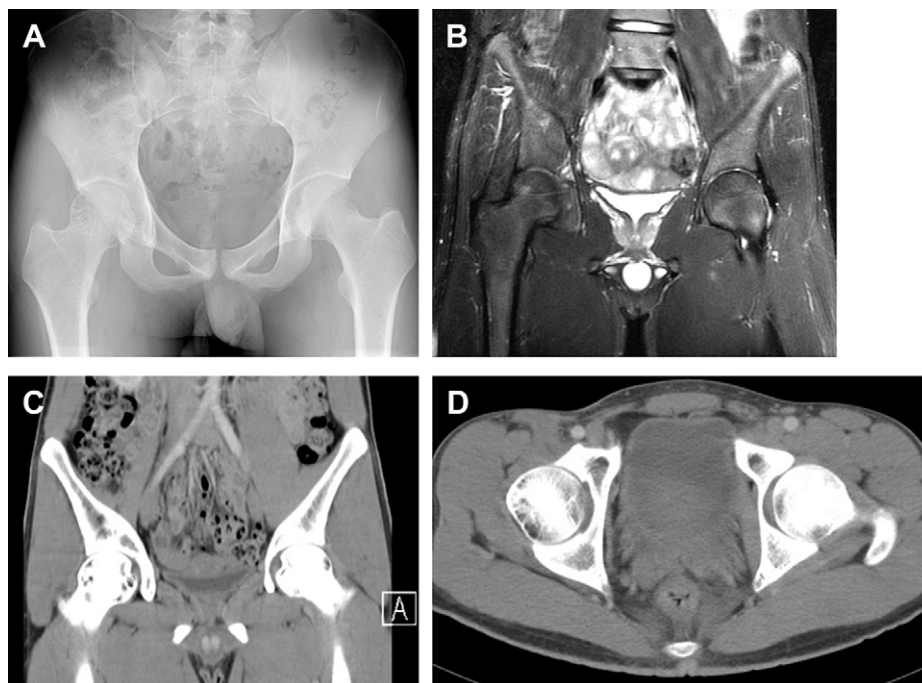


Fig. 3. (A) Anteroposterior radiograph at 3 years postoperatively revealed a well-regenerated acetabular wall. (B) Magnetic resonance imaging, (C) coronal and (D) axial computed tomography of the pelvis revealed no evidence of recurrence and good congruence of the right hip joint.

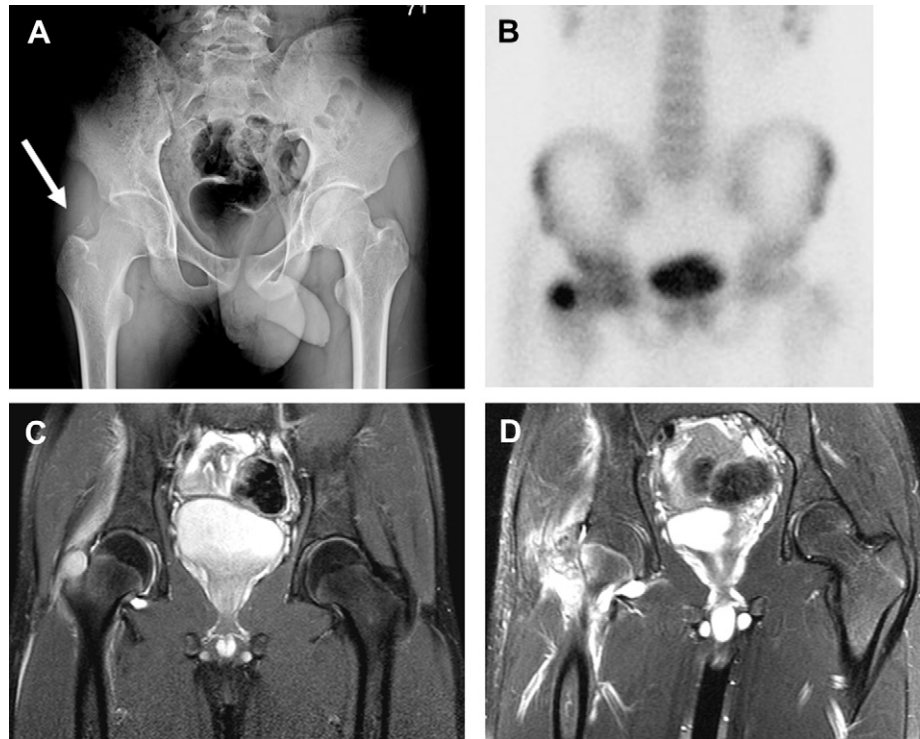


Fig. 4. (A) Anteroposterior radiograph of the pelvis revealed an osteoblastic lesion over the right greater trochanter. (B) Whole-body bone scan showed an intense focus at the right greater trochanter, increased soft-tissue activity in the lateral aspect of the right hip, and diffuse uptake in the right femoral head and neck. (C) Coronal magnetic resonance imaging of the pelvis revealed a tumor growing outward from the right greater trochanter and femoral neck, and neighboring muscle and fascial edema, bone marrow edema at the right femoral neck and head, and hip joint effusion were noted. (D) Diffuse postoperative scarring from a previous lateral approach.

larger at presentation (usually >2 cm in diameter) and exhibit a more prominent periosteal reaction.¹¹ A variant termed “aggressive osteoblastoma”⁷ has similarities to osteosarcoma, radiographically demonstrating cortical destruction and a more prominent periosteal reaction with occasional soft-tissue extension, hence the distinction must be based on histological examination.¹²

For the detection of osteoblastomas, computed tomography (CT) appears to be a more valuable imaging than MRI because it can clearly demonstrate the tumor size, location, central calcification within lesion (typical finding for osteoblastoma), and delineate the cortical destruction or soft-tissue extension.

Generally speaking, MRI would be the choice of imaging study following plain radiograph for early detection of suspected pathological lesions; however, Crim et al¹³ have reported “flare phenomenon”, which is a widespread inflammatory response surrounding an osteoblastoma that leads to diffuse, reactive inflammatory infiltration, thus masking the presence of the tumor. Von Chamier et al¹⁴ reported the case of a 14-year-old male with hip pain whose MRI revealed joint effusion and bone marrow edema of the right femoral neck. Chronic nonbacterial osteomyelitis was diagnosed after repeat biopsy and MRI study. However, the patient’s clinical signs and symptoms were fluctuating, and the

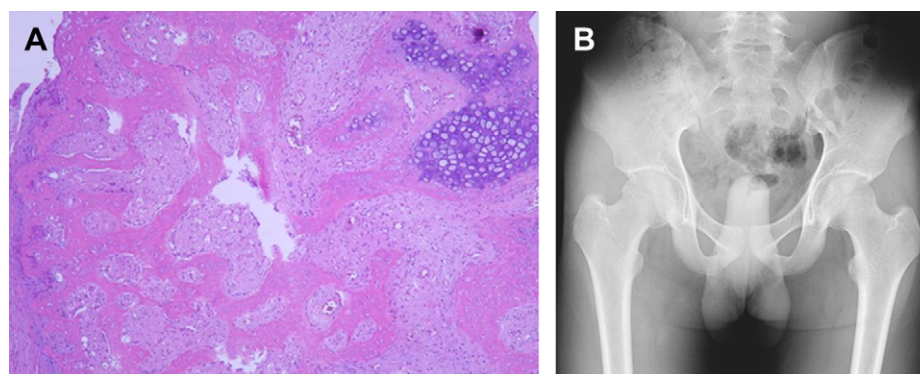


Fig. 5. (A) Histology showed interlacing osteoid trabeculae with osteoblast rimming (hematoxylin and eosin; original magnification $\times 40$). (B) Postoperative 23-month follow-up anteroposterior radiograph of the pelvis revealed well-healed lesion with no evidence of local recurrence.

Table 1
Clinical data of seven patients with osteoblastomas in the region of the hip.

Case	Age	Gender	Duration of symptoms (months)	Symptoms	Initial histological diagnosis	Final histological diagnosis	Location	Size (cm)	Treatment	Recurrence	Follow-up (months)	Remarks
1	13	M	6	Hip pain Night pain RHM	OGS	OB	Acetabulum	6.9 × 6.2 × 5.9	Curettage + phenol	No	38	Free ROM Harris Hip Score: 100
2	14	M	6	Hip pain	Septic arthritis Tenosynovial giant cell tumor	OB	Femoral neck	2 × 2 × 2	Curettage + phenol	No	6	Free ROM Harris Hip Score: 100
3 ⁵	6	M	12	Hip pain RHM	Legg-Calvé-Perthes disease Synovitis	OB	Acetabulum	2.5 × 2 × 0.5	Curettage + BG	No	12	Free ROM Coxa magna
4 ⁶	26	F	12	Thigh pain RHM		OB	Ischio-pubic		Anterior pelvis resection	No	36	Free ROM
5 ⁷	27	F	12	Hip pain		Aggressive OB	Acetabulum	1.5	Curettage + BG Resection + total hip replacement	Yes No	54	
6 ³	18	M	12	Hip pain, RHM Weight loss Diarrhea	Enteropathic arthritis	OB	Acetabulum		Curettage + phenol + BG	No	24	Free ROM
7 ⁸	20	M	36	Low back pain	Sacroiliitis	OB	Sacroiliac joint	5	Curettage	No	24	Free ROM

Note: Cases 1 and 2 are the cases presented in this report. Cases 3, 4, 5, 6, and 7 are summarized from references 5, 6, 7, 3, and 8, respectively. BG = bone graft; OB = osteoblastoma; OGS = osteogenic sarcoma; RHM = restricted hip motion; ROM = range of motion.

diagnosis was finally established after a CT scan showed a nidus within femoral neck. A wide excision was performed, and histological examination of the specimen was consistent with an osteoblastoma.

Because misinterpretation of pathological specimens is not uncommon, repeat biopsy should be considered if symptoms persisted after initial treatment. The typical histological features of osteoblastomas include an interlacing net of osteoid trabeculae, which is made of woven bone surrounded by predominantly osteoblasts and some spindle cells, giant cells, or blood vessels. Therefore, osteoblastomas can mimic other tumors such as a giant cell tumor due to the variable number of giant cells found on the surface of bone trabeculae. In addition to radiographic similarities, the aggressive osteoblastoma also resembles osteosarcoma histologically as it contains large epithelioid-like osteoblasts surrounding an unorganized osteoid matrix and cells having one or more prominent nucleoli.¹² Kenan et al⁷ stated that the important features differentiating osteoblastomas from osteosarcoma are absence of metastases, atypical mitoses or cartilage cells, and a sharp demarcation between the edge of the osteoblastoma and the host lamellar bone.

Wide excision, rather than curettage, is the treatment of choice for osteoblastomas because of a reported recurrence rate of 9.8–24%.^{1,15} However, when osteoblastomas occur in the hip region, size and location are important concerns when considering operative options. Wide excision is suitable for the non-weight-bearing areas involving the iliac wing, pubic rami, or ischium because postoperative functional impairments are limited.³ On the contrary, for osteoblastomas located within weight-bearing areas such as the acetabulum, preserving hip function is a major concern, and bone graft reconstruction may be required.⁸ In Case 1, the inner wall of the acetabulum was aggressively curetted such that the femoral head was almost visible; nevertheless, reconstruction with bone graft was not performed because of the high regenerative capability of a juvenile. Radiographs and CT 3 years after surgery showed a well-regenerated acetabular wall with good congruence and no evidence of recurrence.

In conclusion, osteoblastoma in the region of the hip is rare and easily misdiagnosed, thus leading to improper treatment. For patients with pelvic pain without a clear origin, pelvic CT should be performed in addition to plain radiographs and MRI, which may be affected by the flare phenomenon. Biopsy prior to surgery is important and very useful for making a diagnosis and deciding what type of procedure should be performed. Intralesional curettage and high-speed burring with or without bone grafting may achieve satisfactory results as compared with wide resection for the treatment of lesions involving weight-bearing areas.

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